

letter to editor

An unusual presentation of Hodgkin's lymphoma: Ectopic testicular involvement

To the Editor: Although the vast majority of testicular lymphomas are diffuse large B-cell type, testicular involvement by Hodgkin's lymphoma is extremely uncommon. Involvement of an ectopic testicle by Hodgkin's lymphoma has never been reported in the literature.

A 17-year-old man was admitted to our institution with an eight-month history of fever, weakness, night sweats and weight loss. He had recently developed a right cervical mass. His medical history was not remarkable. At first presentation, his performance status was equal to 1 according to the Eastern Cooperative Oncology Group (ECOG) scale. Physical examination showed a right cervical node of 2×1.5 cm and an empty left scrotum. Laboratory findings on admission showed high erythrocyte

sedimentation rate (86 mm/h). The computed tomography scan revealed right cervical, mediastinal and retroperitoneal lymph node enlargement with multinodular splenomegaly. Ultrasonography showed a heterogeneous intra-abdominal left testicle. Left radical orchiectomy and cervical node biopsy were performed. Histological and immunohistochemical analysis of both locations confirmed the diagnosis of Hodgkin's disease of a nodular sclerosis subtype. Most of the neoplastic cells were positive for CD15 and CD30 (Figure 1). The result of his bone marrow biopsy was normal. His disease was staged at IVB according to the Ann Arbor classification system.

Our patient was treated using eight courses of chemotherapy every four weeks with ABVD regimen: doxorubicin 25 mg/m² on day 1 and 15, bleomycin 15 mg on day 1 and 15, vinblastine 10 mg on day 1 and 15, and dacarbazine 375 mg/m² on day 1 and

15. His radiological evaluation after the eighth cycle of chemotherapy showed complete response. The patient remains in good control after 26 months of follow-up.

Malignant lymphoma of the testis accounts approximately for 5% of all testicular neoplasms and 1% of all lymphomas.¹ It is usually a late manifestation of extended disease. Most cases of testicular lymphoma have diffuse large B-cell histological features.² Rare cases of Burkitt's lymphoma, follicular lymphoma and peripheral T cell lymphoma have also been found in the testis.³

Testicular lymphoma occurs almost exclusively in patients over 60 years of age. It also constitutes, with spermatocytic seminoma and Leydig cell tumors, the most common testicular malignancy in men older than 50 years of age.¹

Testicular involvement is an unusual presentation of Hodgkin's lymphoma. At previous autopsy reports, microscopic testicular infiltration was found in none of

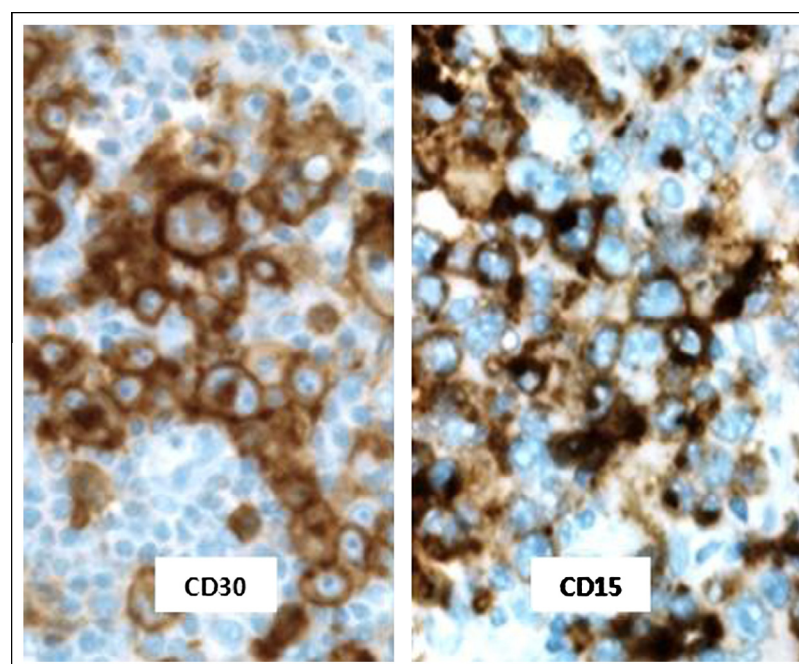


Figure 1. Neoplastic cells are positive for CD30 and for CD15.

Table 1. Clinical characteristics, pathological features and treatment of patients with testicular Hodgkin's lymphoma in the literature.

Author	Age	Clinical information	Site (testis)	HL subtype	IHC	Stage	Treatment	Outcome
Our case	17	Cervical mass Empty scrotum	Left	NS	CD15+ CD30+	IVB	Orchiectomy + CT (8 × ABVD)	CR (DFS 2 years)
Gatt (2011) (1)	73	Testicular mass	Right	NS	CD3+ CD15+ CD30+	IVA	Orchiectomy	Died before CT
Seliem (2007) (5)	52	Testicular mass	Left	Classical	CD15+ CD30+	IVB	Orchiectomy + CT (4 × ABVD and 2 × AVD)	CR
Glaholm (1989) (6)	56	Scrotal pain Axillary node	Right	MC	np	IVA	Orchiectomy + CT (6 × ChIVPP)	CR (DFS 3 years)
Vishniavsky (1973) (7)	79	Inguinal mass	Right	Hodgkin sarcoma (autopsy)	np	—	—	Died before treatment

HL: Hodgkin's lymphoma; IHC: immunohistochemistry; NS: nodular sclerosis; MC: mixed cellularity; np: not performed; CT: chemotherapy; ABVD: doxorubicin, bleomycin, vinblastine, dacarbazine; AVD: doxorubicin, vinblastine, dacarbazine; ChIVPP: chlorambucil, vinblastine procarbazine, prednisolone; CR: complete response; DFS: disease free survival.

the patients screened with Hodgkin's disease.⁴ Also, to our best knowledge, only four cases of Hodgkin's lymphoma with testicular involvement have been reported previously in the English literature^{1,5–7} (Table 1), and confirmation of Hodgkin's disease by immunohistochemistry was obtained in only two cases. This present case is the first one describing Hodgkin's lymphoma in an ectopic testicle.

As primary testicular Hodgkin's lymphoma has not yet been reported, secondary testicular involvement should be considered in case of extensive Hodgkin's disease. Two hypotheses can be suggested to explain testis tissue involvement: directly contiguous spread from adjacent lymph nodes and hematogenous spread.⁵ In our case, the first theory was suspected because of the presence of enlarged lymph nodes adjacent to the intra-abdominal testis.

There is no standard management for malignant lymphoma of

the testis. However, recognition of this rare entity is very important to avoid orchiectomy as lymphomas are systemic diseases and their treatment is based on chemotherapy. Our patient was managed successfully with ABVD chemotherapy and remains disease free two years after completion of treatment.

CONFLICT OF INTEREST

None declared.

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